

Celiac disease and intussusception – a rare but important association

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Abstract

Most of the cases of intussusception in children are labeled as idiopathic and not usually investigated. Only rare cases of chronic or recurrent intussusception receive greater attention and further investigations. Celiac disease appears to be associated with modestly increased risk of intussusception, although most of these cases remain asymptomatic. Debate still continues as to whether all patients of intussusception should be screened for celiac disease. We present case of a 7 years old child who presented with recurrent intussusception, underwent laparotomy initially and was later diagnosed to have celiac disease. (*Acta gastroenterol. belg.*, 2022, 85, 111-112).

Keywords: Celiac disease, gluten-free diet, intussusception.

Introduction

Intussusception is one of the common causes of intestinal obstruction in children in the first five years of age and usually manifests as episodic pain abdomen and rectal bleeding. More than 70% of the cases of intussusception are idiopathic with ileo-colic being the most common site (1). Celiac disease (CD) is an immune mediated disease leading to damage to small intestinal mucosa in genetically predisposed host, precipitated by gluten. Although the prevalence of CD in most regions is < 1%, studies from northern India have reported CD prevalence rates of 10% to 26% in children (2). Clinical manifestations of CD can vary from that of chronic diarrhea and other features of malabsorption to non-classical manifestations. Patients with celiac disease who do not present with the classical symptoms are usually diagnosed late. Patients with CD can develop intussusception but it is mostly asymptomatic. Debate is still on whether all children labeled as idiopathic intussusception should undergo antibody testing for celiac to prevent future episodes of obstruction. We present a case of a child who presented with intestinal obstruction due to intussusception and was diagnosed to have CD.

Case

An 11 year old male child presented to Gastroenterology department with history of pain abdomen and vomiting since 3 days. On examination, the child was dehydrated and had tachycardia. His abdomen was distended with absent bowel sounds. There was history of similar



Figure 1. — Cross-sectional ultrasound image showing intussusception.

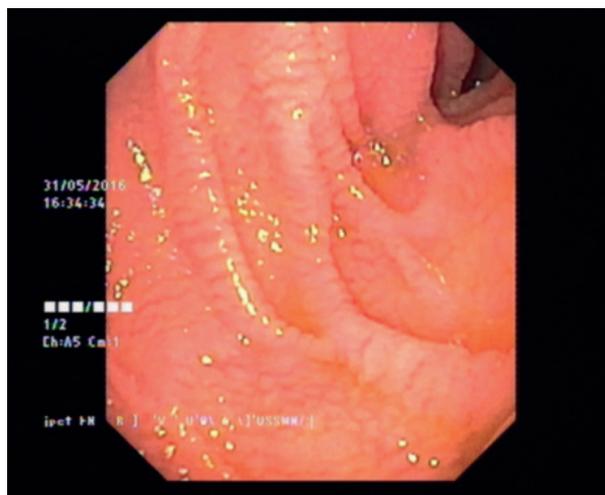


Figure 2. — Endoscopic image of duodenum showing grooving and scalloping of folds.

complaints 8 months back, when he was diagnosed to have small bowel obstruction due to intussusception. He underwent laparotomy at that time at a local hospital. There was also history of failure to gain weight since 2 years. There was no history of loose stools or fever.

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Lab investigations revealed Hb - 7.5 mg%, blood sugar-98 mg/dl, normal renal function tests and normal liver function tests except low serum albumin (2.8 mg/dl).

X Ray abdomen showed multiple air fluid levels suggestive of small gut obstruction. The child was kept nil per oral with gastric aspiration. Ultrasonography of abdomen revealed intussusception of the small bowel. On getting work up of anaemia, serum Iron and ferritin were found to be low and TIBC was high. As CD is common in our region, serum anti-tissue transglutaminase (Anti-TTG) antibody levels were done, which were found to be high (> 300 IU/ml). The child was managed conservatively and there was gradual resolution of obstruction. Upper GI endoscopy revealed scalloping of duodenal folds and duodenal biopsy showed partial villous atrophy. Thus, the diagnosis of celiac disease was made. He was started on gluten free diet and discharged in satisfactory condition. After 6 months of follow-up, the child had gained weight and also showed increase in haemoglobin.

Discussion

Intussusception is one of the most common causes of intestinal obstruction in pediatric age group and it needs prompt intervention (surgical or non surgical). Surgical modalities are used in complicated intussusception like perforation, peritonitis or in cases of shock, while non-operative modalities like pneumatic reduction and enema can be used in most of the other cases. In contrast to adults where intussusception is mostly pathological (3), in children it demands investigations only if examination shows some abnormality.

Intussusception it is probably under diagnosed in celiac patients, as it may be chronic and painless. As celiac is a chronic inflammatory condition, the proposed cause of intussusception in cases of celiac disease is inflammation and thickening of the intestinal wall leading to hyper peristalsis and subsequent dilation of the small bowel. This, in turn, could be the lead point for intussusception, which can develop singly or multiply, chronically and with self-resolution or the need for surgical intervention. Less frequently, but with worse prognosis, this lead point could also be associated with a focal lead point in lymphomas.

Association between CD and intussusception has gained attention in the recent years. However, majority of the data regarding this association is in the form of case reports or small case series. In a recent study

from northern India, Borkar et al prospectively studied the prevalence and natural history of intussusception in newly diagnosed CD. In this study, 25% of the children with newly diagnosed CD had subclinical intussusception (4). However, only one out of 37 children was symptomatic. In another study in children with symptomatic intussusception, the prevalence of CD was 1.2% (5). In a study by Gonda et al, 57% of the adults presenting with intussusception were diagnosed to have celiac disease (6). However, there has been some conflicting data regarding this association. In one of the case control studies done by Ludvigsson et al showed no correlation intussusception and future risk of CD (7).

What makes our case important is the recurrent presentation of intussusception with requirement of laparotomy in a child with growth retardation and anemia. During the current episode, the diagnosis of CD was made and also the intussusception resolved spontaneously with gluten free diet. CD was the etiological factor for intussusception as the child remained asymptomatic after starting gluten free diet.

To conclude, intussusception is a common cause of idiopathic acute intestinal obstruction in children. Celiac disease could be an important etiological factor for idiopathic intussusception in children. Therefore, screening for celiac disease with a non-invasive blood testing should be done in children with intussusception, which can save the child from future risk of complications and surgical intervention.

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